

Modified Marsupialization of Simple Ranula in a Child and Review of Management Techniques

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Abstract:

A ranula is a bluish dome shaped swelling in the floor of the mouth resulting from accumulation of saliva from the sublingual gland. It is of two types simple and plunging ranula. The management is difficult due to its anatomic location. Children pose a greater challenge while managing this condition due to their age and behavioural problems.

An 8-year-old girl reported with a sublingual ranula on the floor of the mouth. The swelling was marsupialized under local anesthesia followed by betadine-soaked gauze packing changed at three-day intervals. Complete resolution of the lesion took place with no signs of recurrence after a follow up of 1 year.

Various treatment options are available for management of pediatric ranulas. Micro-marsupialization and marsupialization are relatively less invasive procedure and its use in management of pediatric ranulas demonstrates lesion resolution with low recurrence when combined with packing of the pseudocyst. Invasive methods like complete excision with or without removal of the sublingual gland can lead to local complications. Other option for management includes lasers, sclerotherapy with OK-432, botulinum toxin type A and nickel gluconate – mercurius heel – potentised swine organ preparations.

Key words: Simple Ranula, Marsupialization, Sublingual Gland

Introduction:

A ranula is seen as a bluish, dome shaped, unilateral, fluctuant, soft, translucent swelling on the floor of the mouth and sometimes extending in the neck, due to accumulation of saliva from the sublingual salivary gland. The pooled saliva initiates an inflammatory response resulting in formation of a pseudocyst^(1,2). The sublingual gland has multiple ductal openings in the floor of the mouth. The largest duct is called 'Bartholin's' ducts while the smaller openings are collectively called as 'ducts of Rivinus'. Salivary extravasation may occur due to trauma, blockage or inflammation of these sublingual gland ducts.

Traumatic injuries, chronic inflammatory diseases and anatomical variations of the sublingual gland are common causes for development of ranulas.^(1,3) Simple ranulas have to be differentiated from enlargements of submandibular gland origin or minor salivary glands that manifest as swellings in the floor of the mouth. Obstruction of the 'Wharton's duct' may lead to sialolithiasis. Injuries to the 'Wharton's duct' cause extravasation and pooling of saliva that may resemble a ranula. Plunging ranulas must be differentiated from swellings that are observed over the neck like thyroglossal duct cyst, dermoid cyst, branchial cyst, lymphangioma, cystic hygroma, laryngocoele, lipoma, arteriovenous

malformations, and enlargements of thyroid or parathyroid glands. The location of the swelling helps to narrow down the list of differential diagnosis, though histopathological confirmation may be required for final diagnosis⁽⁴⁾.

Selection of the surgical technique is important for successful resolution of ranula. It should be based on the size of the lesion, type of ranula, history of previous intervention with recurrence and risk of damage to important anatomical structures. In case of children, the age and behavioural assessment should also be considered to chart the most appropriate plan of treatment.

Case Report:

An eight-year-old girl reported for oral examination with a chief complaint of swelling over the floor of the mouth on the right side since 10 – 12 days (Fig. 1).



The swelling was ovoid in shape approximately 2 X 1 cm in dimension, reddish in colour, soft and fluctuant. Patient informed that the swelling was noticed recently and there was no change in size or history of spontaneous resolution and recurrence. The swelling did not interfere in mastication or speech, it was not painful or tender on palpation. It was not compressible and no discharge was noticed. Relevant haematological investigations were performed and a mandibular occlusal radiograph was taken to rule out obstruction of the Wharton's duct. Based on examination and investigations, a clinical diagnosis of ranula was made. Parents were explained about various treatment modalities while informing about potential recurrence and complications. Parental consent was obtained to perform marsupialization of the lesion.

The patient's behavioural assessment was done and it was decided to perform the procedure under local anesthesia. Extra- and intra-oral betadine scrubbing was done following administration of local anesthesia. Subjective and objective signs of adequate anesthesia were confirmed. An elliptical incision was given at the base of the lesion taking care to limit the depth of the incision to prevent injury to the Wharton's duct and neuro-vascular structures at the base of the tongue (Fig. 2).



The exudation of mucinous fluid following the incision confirmed the presence of saliva. Haemostasis was achieved with pressure packs. The lining of the ranula was identified circumferentially and a continuous mattress suture was taken around the lesion, suturing the lining of the ranula with the oral mucosa using 3-0 polyglycolic acid resorbable material. The defect was packed with betadine-soaked sterile gauze pack (Fig. 3).



The patient was recalled to change the gauze pack at three-day intervals for 3 cycles, after which the packing of the surgical cavity was discontinued. Complete resolution of the wound and resorption of the sutures were seen one month after the surgery and there was no recurrence at one-year follow up (Fig. 4).



Discussion:

Ranula is a fluid filled lesion seen on the floor of the mouth following traumatic injury or obstruction of the sublingual salivary gland duct⁽⁵⁻⁹⁾. The secretion of the sublingual gland is mucinous compared to other salivary glands. Leakage of such viscous fluid generates a strong inflammatory reaction from the surrounding tissues⁽⁵⁾. Secretion of saliva from the sublingual gland is continuous and this prevents the parenchymal tissue of the gland from undergoing inflammatory granular or fibrous metamorphosis. Hence ranulas are commonly associated with the sublingual gland⁽¹⁾. According to its site of presentation and extent, they are classified as sublingual (visible only in the oral cavity below the tongue), sublingual-submandibular (visible in the mouth and also in the submandibular region) and submandibular (visible only in the submandibular region without visible intra oral involvement). The sublingual variant is called as simple ranula while the latter are examples of plunging ranula^(9,10).

Prevalence of ranulas is 0.2 cases per 1000 persons while the prevalence of congenital ranulas in children is 0.74 percent and it is a rare clinical entity. They comprise of about 6 percent of all oral sialoceles⁽¹¹⁾. A simple ranula is common during the first and second decades of life while the plunging ranula is observed during the third decade with a female predilection of about 1.15:1⁽¹²⁾. Ranulas may interfere in mastication and can be painful if inflamed. It also causes difficulty in oral hygiene maintenance. The patients may complain of variance in the size of the swelling or recurrence over a period of time.

Traditional options in management of ranula include incision and drainage, excision of the ranula, with or without removal of the ipsilateral sublingual gland, marsupialization, with or without packing the surgical defect. Newer methods of management include micro-marsupialization, modified micro-marsupialization, lasers and injectable agents⁽¹³⁾. In the

present case the surgical defect was marsupialized and packed with a betadine-soaked gauze. The pack was changed at three-day intervals. Regular changing of the gauze pack allowed for careful monitoring of healing, examining the wound for debris collection while betadine provided antibacterial effect to prevent infection of the surgical wound. Management of ranulas is challenging due to its anatomical location. The floor of the mouth has vital neurovascular structures that may be damaged during surgical exploration. Unrestricted movement of the tongue can lead to post operative bleeding and dehiscence of the surgical wound. Contamination of the wound can cause infections, pain and difficulty in swallowing⁽¹⁴⁾.

The lingual nerve and the submandibular gland duct are at higher risk of injury during surgical management in infants and children, due to their close proximity to the sublingual gland [14]. The common complications of ranula include recurrence (5.78%), paraesthesia of the tongue (4.89%) and damage to the Wharton's duct (1.82%)⁽¹⁵⁾. Resection of the sublingual gland along with excision requires the procedure to be done under general anesthesia. It is an invasive method and has a higher probability for post operative complications⁽¹⁵⁾. Recurrence of ranula has been attributed to the size of the lesion at the time of presentation and the surgical method employed for its management. The recurrence rates for incision and drainage are 71-100%, 61-89% for marsupialization, 0-25% for excision of ranula, 0-12% for marsupialization with packing and 0-2% for excision of ranula with sublingual gland⁽¹⁴⁾.

Review of Management Techniques:

Many surgical methods have been advocated for management of ranulas. Observation for spontaneous resolution up to 6 months after it is observed has been advised in children. Intervention is recommended if there is no resolution or recurrence during the observation period⁽¹⁶⁾. Excision of the ranula, excision with removal of ipsilateral sublingual gland, marsupialization and micro-marsupialization are commonly encountered techniques in the literature. Besides surgical management, CO₂ laser excision and vaporization of non-plunging ranulas have been compared by Mintz et. al⁽¹⁷⁾.

Novel approaches in the form of intra-lesional injection of lyophilized *Streptococcus pyogenes* preparations like OK-432 is under experimental use for the treatment of ranula^(10,18). A recent study has recommended the use of Nickel Gluconate – Mercurius Heel – Potentised Swine Organ Preparation D10/D30/D200, which is a homotoxicological agent as an alternative treatment strategy for ranulas⁽¹⁹⁾.

Excision of the ranula with or without the excision of the sublingual gland:

Radical approach by excision of the ranula along with the sublingual gland had been advocated by Pandit and Park (2002) to reduce the rate of recurrence⁽¹⁶⁾. They suggested that dissection around the submandibular gland duct and relocating it increases the exposure of the floor of the mouth for exploring the boundaries of the excision site⁽¹⁶⁾. Crysedale et al. recommended sublingual gland removal if the ranula was greater than 1 cm⁽²⁰⁾, while Bridger et al.⁽²¹⁾ and Catone et al.⁽²²⁾ advised gland excision as the primary treatment option irrespective of the size of the lesion. An explorative approach in management of ranula carries a potential risk of haemorrhage from the lingual and sublingual vascular structures, paraesthesia of the lingual nerve and damage or repositioning of the Wharton's duct. A large lacrimal probe or catheter inserted in the duct of the submandibular gland at the time of surgery helps to identify its course and prevent injury⁽¹⁴⁾. Yoshimura et al reported the results of 27 cases of plunging ranulas. Four patients had excision of ranula only and 9 patients underwent removal of ranula with sublingual gland they showed a recurrence of 25.0% and no recurrence respectively⁽⁹⁾.

Marsupialization:

Marsupialization of ranula is considered as a less invasive option for management of ranula in pediatric age group. A tiered approach of marsupialization followed by resection in case of recurrence is followed by some clinicians to avoid initial extensive exploration. The entire lesion is unroofed and the lining of the ranula is sutured with the oral epithelium. In an alternative method, Baurmash suggested packing the pseudocyst with gauze for 7 – 10 days. the packing material prevents the margins of the cavity to reunite, allows re-epithelization of the cyst cavity, seals the mucinous leak and incites a foreign body reaction leading to fibrosis and atrophy of the gland acini⁽²³⁾.

Micro-marsupialization:

Placement of a single silk suture over the most prominent part of the ranula to drain the mucinous fluid and maintaining the suture for 10 days allowed for epithelization along the suture tracts. This technique called as micro-marsupialization provides an alternative, conservative approach in children with ranulas⁽²⁴⁾. Delbem et al reported that out of 14 patients treated by micro-marsupialization, 12 presented full lesion resolution after one week while two cases showed signs of recurrence⁽²⁵⁾. In another case multiple silk sutures placed over the length of the swelling and maintained for 30 days to allow for epithelization of the suture tracts has been suggested by Hegde et al.⁽²⁶⁾. They suggest that creating multiple openings allowed for better drainage and reduced risk of recurrence.

Laser excision:

Laser excision and vaporization of ranula with CO₂ and Er,Cr:YSGG lasers have been reported by Frame, Mintz et al and Barak et al^(17,27,28). The laser beam is absorbed by the intracellular water of the oral mucosal cells. The transferred energy results in photo-thermal rupture of the cells. This causes vaporization and sealing of minor salivary gland ducts and acini⁽²⁹⁾. There was no reported recurrence after 6 months in a case series by Mintz et al⁽¹⁷⁾ which was consistent with the reports of Niccoli-Filho and Morosolli who combined marsupialization with CO₂ laser excision⁽³⁰⁾. Zola et al reported management of a 12 year old patient with ranula using Er,Cr:YSGG laser⁽³¹⁾. The use of soft tissue lasers gives a promising alternative to traditional surgical methods to reduce post operative complications in the treatment of ranula.

Sclerotherapy with OK-432:

Injection of a lyophilized mixture of group-A Streptococcus pyogenes having anti neoplastic activity called as OK-432 (Picibanil®) has been experimentally tried for the successful resolution of oral ranulas. It's use in the management of lymphangiomas and cystic hygromas has been well documented, but its effects for ranula management needs further research. Intra cystic injection of OK-432 triggers a host immune response with pain and sclerosis at injection site with fever. It is considered as an alternative line of treatment for patients who do not wish to undergo surgical intervention⁽³²⁾.

Botulinum Toxin type A

Local injection of botulinum toxin type-A causes denervation of the para-sympathetic nerves responsible for salivation. A case series of three patients reported successful treatment of ranula after injection of botulinum toxin type-A⁽³³⁾. The use of botulinum toxin for neuromuscular and cosmetic treatments has provided data on its safety and tolerability. Its use for management of ranulas is still considered as experimental⁽³⁴⁾.

Nickel Gluconate – Mercurius Heel – Potentised Swine Organ Preparations:

Nickel Gluconate is an oxide reducer and stimulates salivary glands in case of lack of amylase deficiency. It stabilizes the cell membrane, stimulates the reticular endothelial system and lymphatic tissue and promotes protein synthesis for tissue repair. Mercurius-Heel is an anti-inflammatory drug and Potentised Swine Organ Preparation, D-10, D-30 and D-200 have specific tropism for homologous human tissue and stimulate protein synthesis. The three homotoxicological drugs used in combination synergistically act on the foreign body inflammation due to extravasated mucus and stimulate glandular repair and pseudocyst reabsorption. In a case series

of 9 children, local injection of this combination drug showed resolution of the ranula in a 4 – 32 month follow up period. Two children showed signs of asthenia and local irritation at injection site due to increase in volume of the fluid. There was no reported allergy or other complications⁽¹⁹⁾.

Conclusion:

Management of ranula requires a planned approach with calculated risk benefit ratio to best suit the needs of the patient and even more so in children due to their delicate physiology and immature psychological and emotional development. A modified marsupialization technique with betadine-soaked gauze packing and regularly changing the pack at short intervals provided successful resolution of the ranula without any post operative complications and no signs of recurrence up to one year of follow up.

References

- Harrison JD. Modern management and pathophysiology of ranula: literature review. *Head Neck*. 2010 Oct;32(10):1310-20. doi: 10.1002/hed.21326. PMID: 20054853.
- McClatchey KD, Appelblatt NH, Zarbo RJ, Merrel DM. Plunging ranula. *Oral Surg Oral Med Oral Pathol*. 1984 Apr;57(4):408-12. doi: 10.1016/0030-4220(84)90160-9. PMID: 6584837.
- Mun SJ, Choi HG, Kim H, Park JH, Jung YH, Sung MW, Kim KH. Ductal variation of the sublingual gland: a predisposing factor for ranula formation. *Head Neck*. 2014 Apr;36(4):540-4. doi: 10.1002/hed.23324. Epub 2013 Jun 1. PMID: 23729331.
- Bhaskar SN, Bolden TE, Weinmann JP. Pathogenesis of mucocoeles. *J Dent Res*. 1956 Dec;35(6):863-74. doi: 10.1177/00220345560350060601. PMID: 13376792.
- Davison MJ, Morton RP, McIvor NP. Plunging ranula: clinical observations. *Head Neck*. 1998 Jan;20(1):63-8. doi: 10.1002/(sici)1097-0347(199801)20:1<63::aid-hed10>3.0.co;2-q. PMID: 9464954.
- Haberal I, Göçmen H, Samim E. Surgical management of pediatric ranula. *Int J Pediatr Otorhinolaryngol*. 2004 Feb;68(2):161-3. doi: 10.1016/j.ijporl.2003.09.017. PMID: 14725982.
- Morita Y, Sato K, Kawana M, Takahasi S, Ikarashi F. Treatment of ranula--excision of the sublingual gland versus marsupialization. *Auris Nasus Larynx*. 2003 Aug;30(3):311-4. doi: 10.1016/s0385-8146(03)00058-0. PMID: 12927300.
- Ghani NA, Ahmad R, Rahman RA, Yunus MR, Putra SP, Ramli R. A retrospective study of ranula in two centres in Malaysia. *J Maxillofac Oral Surg*. 2009 Dec;8(4):316-9. doi: 10.1007/s12663-009-0077-9.

- Epub 2010 Apr 24. PMID: 23139535; PMCID: PMC3454109.
9. Yang Y, Hong K. Surgical results of the intraoral approach for plunging ranula. *Acta Otolaryngol.* 2014 Feb;134(2):201-5. doi: 10.3109/00016489.2013.831481. Epub 2013 Oct 16. PMID: 24128284.
 10. Woo JS, Hwang SJ, Lee HM. Recurrent plunging ranula treated with OK-432. *Eur Arch Otorhinolaryngol.* 2003 Apr;260(4):226-8. doi: 10.1007/s00405-002-0482-x. Epub 2002 Nov 8. PMID: 12709809.
 11. Loney WW Jr, Termini S, Sisto J. Plunging ranula formation as a complication of dental implant surgery: a case report. *J Oral Maxillofac Surg.* 2006 Aug;64(8):1204-8. doi: 10.1016/j.joms.2006.04.032. PMID: 16860210.
 12. Packiri S, Gurunathan D, Selvarasu K. Management of Paediatric Oral Ranula: A Systematic Review. *J Clin Diagn Res.* 2017 Sep;11(9):ZE06-ZE09. doi: 10.7860/JCDR/2017/28498.10622. Epub 2017 Sep 1. PMID: 29207849; PMCID: PMC5713871.
 13. Yoshimura Y, Obara S, Kondoh T, Naitoh S. A comparison of three methods used for treatment of ranula. *J Oral Maxillofac Surg.* 1995 Mar;53(3):280-2; discussion 283. doi: 10.1016/0278-2391(95)90224-4. PMID: 7861278.
 14. Zhao YF, Jia J, Jia Y. Complications associated with surgical management of ranulas. *J Oral Maxillofac Surg.* 2005 Jan;63(1):51-4. doi: 10.1016/j.joms.2004.02.018. PMID: 15635557.
 15. Chung YS, Cho Y, Kim BH. Comparison of outcomes of treatment for ranula: a proportion meta-analysis. *Br J Oral Maxillofac Surg.* 2019 Sep;57(7):620-626. doi: 10.1016/j.bjoms.2019.06.005. Epub 2019 Jun 22. PMID: 31239229.
 16. Pandit RT, Park AH. Management of pediatric ranula. *Otolaryngol Head Neck Surg.* 2002 Jul;127(1):115-8. doi: 10.1067/mhn.2002.126590. PMID: 12161740.
 17. Mintz S, Barak S, Horowitz I. Carbon dioxide laser excision and vaporization of nonplunging ranulas: a comparison of two treatment protocols. *J Oral Maxillofac Surg.* 1994 Apr;52(4):370-2. doi: 10.1016/0278-2391(94)90439-1. PMID: 8133368.
 18. Rho MH, Kim DW, Kwon JS, Lee SW, Sung YS, Song YK, Kim MG, Kim SG. OK-432 sclerotherapy of plunging ranula in 21 patients: it can be a substitute for surgery. *AJNR Am J Neuroradiol.* 2006 May;27(5):1090-5. PMID: 16687549; PMCID: PMC7975734.
 19. Garofalo S, Briganti V, Cavallaro S, Pepe E, Prete M, Suteu L, Tavormina P. Nickel Gluconate-Mercurius Heel-Potentised Swine Organ Preparations: a new therapeutical approach for the primary treatment of pediatric ranula and intraoral mucocele. *Int J Pediatr Otorhinolaryngol.* 2007 Feb;71(2):247-55. doi: 10.1016/j.ijporl.2006.10.013. Epub 2006 Nov 20. PMID: 17116334.
 20. Crysedale WS, Mendelsohn JD, Conley S. Ranulas--mucoceles of the oral cavity: experience in 26 children. *Laryngoscope.* 1988 Mar;98(3):296-8. doi: 10.1288/00005537-198803000-00011. PMID: 3343879.
 21. Bridger AG, Carter P, Bridger GP. Plunging ranula: literature review and report of three cases. *Aust N Z J Surg.* 1989 Dec;59(12):945-8. doi: 10.1111/j.1445-2197.1989.tb07636.x. PMID: 2688626.
 22. Catone GA, Merrill RG, Henny FA. Sublingual gland mucus-escape phenomenon--treatment by excision of sublingual gland. *J Oral Surg.* 1969 Oct;27(10):774-86. PMID: 5258990.
 23. Baumash HD. Marsupialization for treatment of oral ranula: a second look at the procedure. *J Oral Maxillofac Surg.* 1992 Dec;50(12):1274-9. doi: 10.1016/0278-2391(92)90226-p. PMID: 1447605.
 24. Morton RP, Bartley JR. Simple sublingual ranulas: pathogenesis and management. *J Otolaryngol.* 1995 Aug;24(4):253-4. PMID: 8551539.
 25. Delbem AC, Cunha RF, Vieira AE, Ribeiro LL. Treatment of mucus retention phenomena in children by the micro-marsupialization technique: case reports. *Pediatr Dent.* 2000 Mar-Apr;22(2):155-8. PMID: 10769864.
 26. Hegde, Sapna; Bubna, Ketan; Rao, Dinesh (2017). Management of Ranula in a Child by Modified Micro-Marsupialization Technique: A Case Report. *Journal of Clinical Pediatric Dentistry*, 41(4), 305–307. doi:10.17796/1053-4628-41.4.305
 27. Frame JW. Removal of oral soft tissue pathology with the CO2 laser. *J Oral Maxillofac Surg.* 1985 Nov;43(11):850-5. doi: 10.1016/0278-2391(85)90221-6. PMID: 3932616.
 28. Barak S, Horowitz I, Katz J, Kaplan I. Experiences with the CO 2 laser in the surgical treatment of intraoral salivary gland pathology. *J Clin Laser Med Surg.* 1991 Aug;9(4):295-9. doi: 10.1089/clm.1991.9.295. PMID: 10149469.
 29. RizoIU IM, Eversole LR, Kimmel AI. Effects of an erbium, chromium: yttrium, scandium, gallium, garnet

- laser on mucocutaneous soft tissues. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1996 Oct;82(4):386-95. doi: 10.1016/s1079-2104(96)80302-7. PMID: 8899775.
30. Niccoli-Filho W, Morosolli AR. Surgical treatment of ranula with carbon dioxide laser radiation. *Lasers Med Sci.* 2004;19(1):12-4. doi: 10.1007/s10103-004-0293-y. Epub 2004 Apr 2. PMID: 15316853.
 31. Zola M, Rosenberg D, Anakwa K. Treatment of a ranula using an Er,Cr:YSGG laser. *J Oral Maxillofac Surg.* 2006 May; 64(5): 823-7. doi: 10.1016/j.joms.2005.11.062. PMID: 16631492.
 32. Fukase S, Ohta N, Inamura K, Aoyagi M. Treatment of ranula with intracystic injection of the streptococcal preparation OK-432. *Ann Otol Rhinol Laryngol.* 2003 Mar; 112(3): 214-20. doi: 10.1177 / 000348940311200304. PMID: 12656411.
 33. Chow TL, Chan SW, Lam SH. Ranula successfully treated by botulinum toxin type A: report of 3 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2008 Jan;105(1):41-2. doi: 10.1016/j.tripleo.2007.04.007. Epub 2007 Aug 30. PMID: 17764989.
 34. Blitzler A, Sulica L. Botulinum toxin: basic science and clinical uses in otolaryngology. *Laryngoscope.* 2001 Feb;111(2):218-26. doi: 10.1097/00005537-200102000-00006. PMID: 11210864.